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**Communication Intervention for Individuals with Down Syndrome: Systematic Review
and Meta-Analysis**

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Abstract

A systematic review was conducted to identify effective intervention strategies for communication in individuals with Down syndrome. We updated and extended previous reviews by examining: (1) participant characteristics; (2) study characteristics; (3) characteristics of effective interventions (e.g., strategies and intensity); (4) whether interventions are tailored to the Down syndrome behavior phenotype; and (5) the effectiveness (i.e., percentage non-overlapping data and Cohen's d) of interventions. Thirty-seven studies met inclusion criteria. The majority of studies used behaviour analytic strategies and produced moderate gains in communication targets. Few interventions were tailored to the needs of the Down syndrome behaviour phenotype. The results suggest that behaviour analytic strategies are a promising approach and future research should focus on replicating the effects of these interventions with greater methodological rigor.

Keywords: Down syndrome, communication, language, meta-analysis, intervention, intensity

Communication Intervention for Individuals with Down Syndrome: A Systematic Review and Meta-Analysis

Individuals with Down syndrome show an early developing pattern of strengths and weaknesses¹, termed the Down syndrome behavioural phenotype, with delays and differences in speech and language development that include vocal imitation, requesting, first words, vocabulary growth, and mastery of grammar^{2,3}. Communication impairments begin in infancy and continue into adulthood, impacting all aspects of life, including education, employment, family, and community. The extensive communication impairments mean individuals with Down syndrome may be unable to appropriately get their basic needs met, ask for help, or even engage in conversation with peers.

Given that language and communication is such a significant area of impairment there is clearly a need for intervention to address the range of communication difficulties. The examination and dissemination, however, of effective interventions for individuals with Down syndrome is limited. Over the last decade, researchers have continued to call for the development and examination of interventions to address the language and communication impairments in individuals with Down syndrome^{4,5}.

Early reviews of interventions for individuals with Down syndrome suggest that early intervention programs resulted in little success or were inconclusive⁶⁻⁸ in improving communication for individuals with Down syndrome. Since then, The New York State Department of Health (NYSDOH)⁹ conducted a review to identify effective intervention strategies for individuals with Down syndrome. They examined intervention literature for individuals with Down syndrome including single-subject and group studies of interventions. Reviewers categorized interventions by strength of evidence. Interventions having two or more

high quality studies supporting their use were considered to have *strong evidence* and interventions with one moderate quality study supporting their use were considered to have *limited evidence*.

The New York State Department of Health Guidelines recommended that interventions for communication and language use the principles of applied behaviour analysis with intervention involving prompting and positive reinforcement, frequent and rapid delivery, with multiple opportunities to practice a skill. The recommendations for behaviour analytic interventions were, however, based on limited evidence largely from one or two moderate quality studies. From this review it was apparent behaviour analysis is a promising approach to addressing communication in individuals with Down syndrome, but that more research was needed. The recommended intensity of intervention (multiple and rapid opportunities) is also consistent with some of the early research on interventions for young children with Down syndrome¹⁰ and supported by more recent work from Warren, Yoder and colleagues^{11–13}.

The NYSDOH review and the reviews^{6–8} before it present several limitations. Reviewers only reported summary ratings for particular interventions strategies (*strong to limited evidence*), but did not report characteristics of individual studies. Detailed participant characteristics were not included, nor was detailed study information (e.g., design, interobserver agreement, and integrity). Further, even though there is a body of research that evaluates interventions for communication among individuals with Down syndrome, no reviews have applied meta-analytic techniques to summarize the existing research on the effectiveness of these interventions.

Since the NYSDOH review, there is also a growing body of literature describing patterns of development in individuals with Down syndrome. The unique profile of individuals with Down syndrome suggest that they may respond differently to intervention than differing

etiologies of disability¹³. As a result researchers have begun to explore ways to tailor interventions to the Down syndrome behavioral phenotype^{13,14}. Within this framework, interventions are designed to address the areas that are demonstrated weaknesses in individuals with Down syndrome while capitalizing on characteristic strengths. For example, an intervention for communication may capitalize on the relative visual processing strength by using visual-vocal instruction to teach language. Delivered early, interventions targeting the phenotypic weaknesses in Down syndrome can minimize potential collateral effects. For example, interventions addressing early motivational issues in Down syndrome could lead to accessing a greater number of challenging skill building opportunities. Fidler (2005)¹ suggests that outcomes for children with Down syndrome may be improved if interventions account for characteristics within the behavioral phenotype.

In light of this emerging literature on the Down syndrome behavioral phenotype, it is important to update the NYSDOH review to identify effective intervention strategies for communication in individuals with Down syndrome. The present review is a systematic analysis of communication intervention studies for individuals with Down syndrome. We aim to update and extend previous reviews by examining: (1) participant characteristics; (2) study characteristics; (3) characteristics of effective interventions (e.g., strategies and intensity); (4) whether interventions are tailored to the Down syndrome behavior phenotype; and (5) the effectiveness (i.e., percentage non-overlapping data and effect size) of interventions.

Methods

Study Identification and Selection

Selection of studies began with a search conducted on September 30, 2014 of the PsycInfo and ERIC databases for English-language studies published between 1967 and 2014 in

order to capture psychological and educational interventions for communication in Down syndrome. Search criteria entered included three groups of keywords. The first group of keywords were synonymous with Down syndrome including: *Down* syndrome*, and *trisomy 21*. The second group of keywords were terms used to describe language and communication and included: *communication*, *language*, and *speech*. The third group of key words were synonymous with intervention and included: *intervention*, *acquisition*, *treatment*, and *teaching*. The first author then used the conjunctions “OR” to combine keywords within a group and the conjunction “AND” to combine groups of keywords to search each possible combination of keywords. Based on the results of the search, articles were identified that met the inclusion criteria described next. Each identified article's reference section was then systematically analyzed for additional studies.

A study was included in the initial collection based on four criteria. First, at least one participant had a diagnosis of Down syndrome. If the article included multiple participants, only those individuals diagnosed with Down syndrome were included in the review. Second, only articles that reported the results of experimental designs (controlled trials, single-case designs) were selected; articles using non-experimental designs were excluded. Third, single-case studies were selected if baseline and treatment phases were present in the study and if repeated data points, not mean scores, were reported. Studies containing less than two baseline data points were excluded for data analysis purposes. Fourth, studies were included if treatment targeted outcomes related to speech, expressive phonology, syntax, or vocabulary. On the basis of title and abstract, the author retrieved 101 studies for detailed evaluation; an additional 24 articles were obtained from the review of reference lists. Potential studies were evaluated against the inclusion criteria, resulting in 37 studies that met inclusion criteria. The screening results from

the first author were compared to those of a second coder (a research assistant) using the same decision criteria. Agreement was strong at 90%.

Variables Coded and Reliability

Participant characteristics. The following participant information was coded: the number of participants with Down syndrome, age, gender, and level of intellectual functioning as reported in the study.

Study characteristics. The following study characteristics were coded: design, evaluation of treatment integrity data and examination of generalization and follow-up.

Intervention characteristics. The following intervention information was coded: opportunity type, intervention strategies, and intensity. Opportunity type was coded as either learner-directed, teacher-led, peer-led or a combination of two or more. Teacher-led interventions refer to those in which learning opportunities were structured, and an adult (interventionist, practitioner, or other individual) directed and initiated opportunities. Peer-led interventions were those in which learning opportunities were directed and initiated by an individual (with or without a disability) identified as a peer of the participant. Learner-directed interventions were those in which opportunities were initiated by the individual with Down syndrome.

Interventions strategies were recorded as indicated by the authors and included: Prompting, reinforcement, naturalistic language paradigm (NLP), milieu teaching, manualized reading and language intervention, and speech recasting.

The following intensity variables were coded: session duration, number of opportunities per session, sessions per week, and the total duration of intervention (in sessions)¹⁵. If intensity

variables differed for individual participants, intensity variables were averaged across all participants with Down syndrome to yield a single value per article.

Tailoring to the Down syndrome behavior phenotype. We determined whether interventions and target behaviours were tailored to the needs of individuals with Down syndrome if the interventions had three characteristics: (1) authors identified the needs of Down syndrome in their rationale, by using the words Down syndrome in the introduction, (2) participants all had a diagnosis of Down syndrome, and (3) the target of intervention was a need identified in the Down syndrome behaviour phenotype as described by Chapman and Hesketh³.

In describing the outcome variables we determined the modality (sign language, AAC devices, or vocalizations), the domain (receptive, receptive/expressive), and the verbal operant (undefined, combination of two or more, echoic, mand, tact, or intraverbal). We coded modality or form of communication as vocalization, sign language, AAC (not including signs), or combinations of two or more modalities. Target behaviours were coded as either a combination of receptive and expressive or expressive only. Expressive target behaviours were spoken responses (or adapted for use with alternative communication systems) by the participant. Receptive target behaviours were nonvocal responses to a teacher's spoken instructions. We also coded the function of the communicative behavior. One way of categorizing the function of communication is Skinner's (1957) verbal behavior model. Instructional targets are described as echoics, mands, tacts, or intraverbals. An *echoic*¹⁶ occurs in response to other verbal behavior, but the resulting verbal behavior matches the form of the verbal stimulus; this is often referred to as verbal imitation. For example, imitation of sounds, words, or entire phrases would be considered echoics. A *mand*¹⁶ is occurs when the response is under the functional control of deprivation or aversive stimulation and reinforced by a characteristic consequence. This is

commonly referred to as a request. Examples of mands include requesting food and toys or asking for a break. A *tact*¹⁶ is evoked by “a particular object or event or property of an object or event” (p. 82). This operant is commonly referred to as a label. *Intraverbal* refers to verbal behavior that is produced in response to other verbal behavior but is not similar in form to the preceding vocalization. A common example of an intraverbal would be answering a question such as “Where do you live?” Verbal operants could also be coded as a combination of two or more, echoic, mand, tact, or intraverbal as indicated by the authors of the study. If the authors did not indicate the verbal operant, reviewers used the descriptions of the target responses to determine the verbal operant. In some cases, not enough information about the target responses was provided to determine the verbal operant resulting in a code of undefined.

Effectiveness of interventions. Percentage of nonoverlapping data (PND)¹⁷ summarizes single-subject treatment efficacy by calculating the percentage of treatment data points that do not overlap with highest or lowest baseline data point. When more than one behaviour was targeted for a participant, the average effect size was calculated by weighting each behaviour according to the number of data points reporting on the behaviour. Within each article effect sizes were weighted according to the number of data points per participant with Down syndrome, then averaged for all participants with Down syndrome to yield a single effect size per article. All effect sizes were calculated by comparing the first baseline phase to the final treatment phase as this method accounts for the potential for researchers to withdraw their first treatment phase prior to achieving maximum effects and issues in return to baseline in reversal designs¹⁸. For alternating treatment designs, the research question for this review concerned the effect of treatment compared to baseline conditions, therefore PND was computed by comparing data in the baseline phase to data under treatment conditions during the alternating treatments phase¹⁸.

PNDs greater than 70% were considered effective interventions, between 50% and 70%, questionable, and less than 50%, not effective^{17,19}.

For group designs, reported Cohen's *d* effect sizes were used, or effect sizes were calculated for posttest group differences from published data. Only data where the results for individuals were reported separately were included. When a study had more than one outcome variable the average of all outcome measures was calculated. Standard interpretation of Cohen's d^{20} was used to consider the strength of evidence for group design studies (i.e., $< 0.2 =$ trivial; $0.2 - 0.5 =$ small; $0.5 - 0.8 =$ moderate; $> 0.8 =$ strong).

Interobserver Agreement. A subset of studies (32.4%) was coded by two raters to permit the calculation of reliability estimates of variables listed in Tables 1 and 2. The second rater held an undergraduate degree in Psychology. Interrater reliability was calculated via point-to-point agreement and was 92.9% (range 81.8% to 100%).

Two raters also calculated PNDs for the same 32.4% of studies. For PNDs, interrater agreement was calculated using the following formula for each study: lowest PND divided by highest PND and multiplied by 100. Interrater agreement for PNDs was 100%.

Results

Thirty-seven articles representing 225 participants with Down syndrome met selection criteria and were included in the analyses (Table 1). Detailed information about the articles is presented in Tables 2 and 3.

Participant Characteristics

Participant characteristics for each study are presented in Table 2. Of the 225 participants, 94 (41.8 %) were male, 69 (30.7 %) were female; gender was not reported for 62 (27.6 %). Individuals ranged in age from 0.8 to 54 years, with a mean of 10.33 years ($SD =$

12.07). IQ was reported for 119 participants, and the average IQ of participants was 50.84 ($SD = 19.74$, $range = 18$ to 87.3). Of those whose IQ was reported, the majority of participants were functioning in the mild range of intellectual disability (67.2 %), followed by moderate (21.8 %), severe (5.9 %), and no intellectual disability (4.2 %).

Study Characteristics

Methodological components of studies are presented in Table 2. Studies were primarily single subject designs: 22 (59.4 %) multiple baseline, four (10.8 %) alternating treatment, four (10.8 %) reversal, and one (2.7 %) multiple baseline and alternating treatment design. Six studies were randomized-controlled trials (16.2 %). While multiple baseline designs are experimental, 5 studies used a multiple baselines across participants design including only 1 participant with Down syndrome and, thus, lacked experimental control for the population of interest.

Generalization data were present in 17 (45.9 %) of the studies. Follow up data were collected in 15 (40.5 %) of studies with a mean follow-up interval of 70.68 days ($SD = 81.58$). Thirty-four studies (91.8 %) collected interobserver agreement, but only 13 (35.18 %) studies collected a measure of intervention integrity.

Intervention Characteristics

Opportunities were teacher-led in 22 (59.4 %) studies, learner-directed in 12 (32.4 %) studies, and peer-led in 2 studies (5.4 %). One study used a combination of learner- and teacher-led opportunities (2.7 %). Twenty-seven (72.9 %) studies used behaviour analytic interventions (prompting or reinforcement) and seven studies (18.9 %) used more naturalistic intervention strategies which included behaviour analytic strategies (five studies [13.5 %] used Milieu teaching, one study used mirroring and responding [2.7 %] and one [2.7 %] study used NLP).

Only two (5.4 %) studies used other interventions which included a manualized reading and language intervention and a morphosyntactic grammar intervention.

Table 4 presents the percentage of studies reporting intensity variables by intervention strategy and Table 5 presents a summary of the intensity characteristics of the interventions. A greater percentage of studies with teacher-led opportunities reported opportunities per session (50 %), sessions per week (86 %) and total intervention duration (91 %), than studies with child-directed opportunities (17 %, 67 % and 83 %, respectively). A greater percentage of studies with child-directed opportunities reported session duration (100 %) than teacher-led intervention studies (68 %). On average, intervention was intense, with sessions occurring daily, lasting for 23.3 minutes, and consisting of just under 20 opportunities.

Tailoring to Down Syndrome Behaviour Phenotype

Table 3 shows the target behaviours and modality for each study. Thirty-six studies targeted expressive behaviour and one study targeted receptive behaviour in addition to expressive behaviour. Twenty-three (62.1 %) studies targeted vocalizations, four studies (10.8 %) targeted sign language, six studies (16.2 %) targeted use of an AAC device, and five studies (13.5 %) targeted a combination of two or more modalities. Verbal operant information was available for 32 studies, with nine (24.3 %) targeting mands, seven (18.9 %) targeting tacts, four (10.8 %) targeting intraverbals, one (2.7 %) targeting echoic, and 11 (29.7 %) targeting two or more verbal operants.

Only 12 of the 37 studies (32.5 %) met all three criteria for tailoring intervention and targets to the Down syndrome behaviour phenotype (Table 3). The majority of studies (78.3%) targeted behaviours identified as areas of critical weakness according to the Down syndrome behaviour phenotype. Fewer studies identified Down syndrome in their rationale (17 studies,

45.9 %) or involved a homogenous population of individuals with Down syndrome (16 studies, 43.2 %).

Effectiveness of Interventions

Means and standard deviations for each effect size grouped according to intervention strategy are shown in Table 4. On average, intervention increased communication targets by 76 %, and, in 13 (35.1 %) of the studies, PND was at least 90 %. This level of PND suggests that intervention for expressive communication for individuals with Down syndrome is effective. Three group studies provided enough information for the calculation of effect size (Cohen's *d*): Burgoyne et al.²¹, Fey et al.¹¹ and Sepúlveda, López-Villaseñor, and Garayzábal Heinze²². Effect sizes were 0.05 (95% CI = [-0.49, 0.58]), 0.3 (95% CI = [-0.48, 1.07]) and 0.75 (95% CI = [-0.19, 1.63]), respectively. Yoder et al.²³ did not report information that permitted calculation of Cohen's *d*, but reported a moderate effect size ($g = .55$) for post-group differences in the number of words spoken after 9 months of milieu communication training. Thus, effect sizes for group studies ranged from trivial to moderate.

Discussion

The overall appreciation of empirically supported interventions among individuals with Down syndrome has been limited by the lack of a synthesis of the available evidence. In order to address this knowledge gap, we conducted a systematic review to examine: (1) participant characteristics; (2) study characteristics; (3) characteristics of effective interventions (e.g., strategies and intensity); (4) whether interventions are tailored to the Down syndrome behavior phenotype; and (5) the effectiveness (i.e., percentage non-overlapping data and effect size) of interventions. A systematic search identified 37 studies. The results of the studies in this review

were largely positive, and included behavior analytic strategies (prompting and reinforcement) for increasing communication, supporting the recommendations of the NYSDOH guidelines.

Participant Characteristics

While the results of this review suggest the potential value of behavior-analytic strategies in increasing communication for individuals with Down syndrome, this conclusion should be considered in light of several aspects of the literature base. Published literature includes a broad age range (0.8 to 54 years), but for the most part, studies are targeting communication in children and youth with Down syndrome. While targeting communication early in life has the potential to minimize later deficits, there would seem to be value in evaluating whether older individuals could be taught additional communication skills to support activities of daily life. For example, as individuals with Down syndrome age, they continue to experience deficits in intelligibility³.

Many studies also included a heterogeneous group of participants, typically a small number of individuals with Down syndrome among a larger group of individuals with intellectual disability. The research in these studies, therefore, did not focus on Down syndrome, rather on the effects of a particular intervention on a particular behaviour. The result of research involving a heterogeneous group of participants is that there is no opportunity to examine intervention developed specifically for individuals with Down syndrome. Given the potential for moderating effects of etiology on intervention outcome¹³, there is a need to investigate interventions designed to specifically address the areas outlined in the Down syndrome behaviour phenotype¹. The only way that these approaches can be validated is by designing studies that focus specifically on individuals with Down syndrome and address targets identified by the Down syndrome behaviour phenotype.

Study Characteristics

Our inclusion criteria yielded a description of the state of the research as it currently exists, allowing commentary on weaknesses in research on interventions for communication for individuals with Down syndrome. Six studies²⁶⁻³⁰ included all four quality indicators (maintenance, generalization, integrity, and reliability) and all had high PND values (ranging from 76% to 100%). Other studies contained an array of methodological inadequacies. General quality standards²⁵ of intervention studies, including maintenance, generalization, and intervention integrity were inconsistently observed. Several investigations also used multiple baseline designs across participants, but only included one participant with Down syndrome. For these studies, experimental control was not demonstrated within the population included in this review, and the results of these designs are subject to the same threats to internal validity as AB designs. As additional quality research studies are conducted, an alternative approach to review could be to conduct a best evidence synthesis³¹, which would have retained only highly rigorous studies.

Intervention Characteristics

With the exception of two studies that were not behaviour analytic and several studies that used more naturalistic interventions with behaviour analytic strategies, studies in this review used behaviour analytic approaches (e.g., prompting and reinforcement). Favorable effects were seen across all behaviour analytic approaches, supporting the recommendations that behaviour analytic approaches are considered best practice in increasing communication in children with Down syndrome⁹.

Despite findings of this and previous reviews, a behavioural approach to intervention has not been widely promoted for individuals with Down syndrome. Recent reviews of speech

impairments in individuals with Down syndrome suggest the need for interventions which target communication, but do not acknowledge the existence of behaviour-analytic interventions^{5,32,33}. The neglect of this approach is so far-reaching in both research and practice that Buckley³⁴ even calls for a “revival” of intensive, behavioural approaches, specifically for individuals with Down syndrome.

The New York State Department of Health Guidelines not only suggest a behaviour analytic approach to intervention but characterized the interventions as relatively intense –a large number of opportunities presented rapidly during frequent session. Overall, interventions appear to be delivered at this sort of high intensity. Interventions in this review were delivered daily, with opportunities provided once every minute, on average. Given that the majority of interventions included within this review are behaviour analytic, this is unsurprising. These interventions tend to involve multiple, teacher-led opportunities presented in close proximity with specific prompting procedures, high rates of reinforcement, and error correction procedures.

In order to account for how variations in intervention intensity impact outcomes, future studies need to report intensity characteristics in greater detail. Many studies report the total number sessions (total intervention duration) delivered to participants and the duration of those sessions, but fewer report how frequently sessions occur or the number of opportunities provided to participants within sessions. Less than a third of the studies report all of these characteristics. It is necessary for researchers to report multiple aspects of intensity so that future analyses can begin to consider dose-response relationships in communication intervention.

Some differences in reporting intensity characteristics may be a function of the different intervention strategies. Different approaches to treatment emphasize intensity differently. For example, interventions with child-directed opportunities may not report, or even have knowledge

of the number of opportunities being presented. Thus, what exactly constitutes a *dose* for these interventions is unclear. Researchers need to consider how interventions in their field could be examined to permit identification of the active ingredients in order to recommend an optimal dose of those interventions. Researchers and clinicians require this information to deliver the active ingredients in the best possible form.

Tailoring to Down Syndrome Behaviour Phenotype

In this review, the studies targeted areas of critical weaknesses, but did not consider the unique contributions of Down syndrome in their rationale or examine the effects of intervention specifically for a population of individuals with Down syndrome. Although many studies targeted areas of critical weakness in young children, this does not indicate that researchers considered the unique needs of individuals with Down syndrome given the criteria used for categorizing areas for young children was quite broad. Interventions were considered to target an area of critical weakness if they targeted expressive vocabulary development in individuals younger than 12. Given that the focus of this review was on studies of expressive communication, any interventions delivered to young children met this criteria. Few studies focused on the more idiosyncratic needs of individuals with Down syndrome, such as grammar development in older childhood and nonverbal requesting in infancy.

Although twelve studies met criteria for tailoring to the Down syndrome behavioral phenotype, they did not show effect sizes larger than interventions which did not. Much of the descriptive literature on Down syndrome suggests that individuals with Down syndrome would benefit from interventions that are tailored to their critical areas of strengths and weaknesses^{1,24} and these ideas need empirical support. For example, Bauer and Jones (2014) created an intervention to teach early requesting skills using the behavioral phenotype to inform

intervention characteristics. The authors targeted a skill which was significantly delayed in children with Down syndrome, provided high rates of social reinforcement (building on the social strength characteristic of individuals with Down syndrome), and taught the skill by breaking it down in to smaller components. In order to deconstruct the requesting skill, intervention progressed through a sequence of requesting situations building from more social (involving attaining caregiver interaction) to more instrumental (involving attaining an object). This sequence was informed by Fidler et al.'s (2005) finding that instrumental requests are more significantly impaired than social requests in infants with Down syndrome. As a result, the intervention improved requesting for three children with Down syndrome.

Effectiveness of Interventions

Our effectiveness calculations showed that most studies resulted in gains in communication for most participants. Prompting and reinforcement, naturalistic language paradigm and milieu teaching all demonstrated PNDs consistent with effective interventions (>70%). Despite differing names, naturalistic language paradigm and milieu teaching also involve behavior analytic techniques (prompting and reinforcement) within the intervention. They differ from highly structured approaches (e.g., discrete trial teaching) by teaching skills in a loosely structured format, where learning trials are initiated by the learner, and by teaching in environments which resemble the typical daily activities that a young child may encounter. If we consider these approaches under the umbrella of behavior analysis, it is clear that behavior-analytic strategies are effective interventions for communication among individuals with Down syndrome.

Close inspection of the data reported in Table 2, indicates that about five of the studies have PND values which rank in the ineffective range according to the criteria by Scruggs et al.

(1987)³⁵. One of these studies used mirroring and responding by peers, where typically developing siblings were taught to imitate what their siblings with Down syndrome said, and respond to vocalizations. The low PND suggests this strategy is less effective than interventions which include prompting and reinforcement.

The other three studies with low PNDs did, however, include prompting and reinforcement, raising the possibility of individual difference variables being responsible for these differential outcomes. In these three studies the individuals with Down syndrome showed severe and profound intellectual disability^{36,37}, limited vocalizations³⁸, and dual diagnosis with autism spectrum disorders³⁹. Individuals with Down syndrome present with heterogeneous deficits and it is likely that these individual differences are also reflected in the included studies. These differences also may have contributed to the range of communication outcomes observed across included studies.

Percentage of non-overlapping data is a commonly used method and one of the most accepted methods to conduct meta-analyses with single-subject research. Although meta-analytic techniques provide unbiased evaluation of the data contained within the studies, the techniques used in this review have limitations. PND provides a metric of effectiveness of intervention, but interpretation of the magnitude of effect based solely on the mean percentage non-overlap is not recommended as PND is not considered a true effect size⁴⁰. Readers should also note the variability in percentage of non-overlapping data for the studies included in this review. Since we did not include “grey” literature, it is possible that the effect sizes in this study are over-estimates of the true effect of intervention. Finally, despite including several randomized controlled trials in this review, only three reported enough data to permit the calculation of an effect size.

Recommendations for Research and Practice

From this review, we can make a number of recommendations for researchers and practitioners considering interventions for communication among individuals with Down syndrome. First, we need studies that focus on the specific needs of individuals with Down syndrome, tailoring intervention to the critical areas of weakness. This also means designing studies that include only participants with Down syndrome. With the growing literature on the behavioral characteristics of individuals with Down syndrome, studies should be designed with the Down syndrome behavioural phenotype in mind. We also need to produce these studies with methodological rigor. Researchers need to monitor the degree to which therapists adhere to treatment protocols as well as maintenance and generalization outcomes. A wide adoption of these standards may establish a clearer picture of the promising effects of behaviour analytic intervention and may constitute the basis for decision-making in public health and social policies relating to Down syndrome and developmental disabilities.

Researchers and practitioners should consider adopting behaviour analytic treatment strategies. This review suggests that there is evidence that behaviour analytic techniques improve communication and language outcomes among individuals with Down syndrome and that behaviour analytic interventions should be recommended for individuals with Down syndrome over other approaches without support. That is not to say that other approaches to intervention are not without merit, merely that we should use them with caution until they have been evaluated by objective methods.

There are many reasons why alternative approaches to intervention have not been sufficiently evaluated. Over many years, practitioners have used commonly accepted interventions for increasing communication in individuals with Down syndrome. Because these

methods are so well established in practice, it is difficult to evaluate them by objective methods. Families and practitioners may be reluctant to participate in trials of already “established” interventions. Other problems in evaluating effectiveness comes from the diversity of individuals with Down syndrome; age, degree of impairment, and associated medical diagnoses are all variables that need to be considered. Evaluations of these interventions need to account for differences other than the just the intervention being investigated. There is a need for researchers with both funding and technical skills to evaluate interventions for communication in Down syndrome in order to prevent practitioners and families from continuing to use interventions without adequate support for their usefulness.

Last, more research is needed to determine the moderating effects of intervention intensity. In order to determine how participant characteristics and other intervention characteristics interact with intervention intensity, there need to be studies with clear descriptions of intensity characteristics. Reporting intensity characteristics allows other researchers to replicate, and allows practitioners to understand the necessity of these characteristics in producing meaningful outcomes for individuals with Down syndrome. This in turn can help inform policies about which interventions and how much of them should be delivered to families of individuals with Down syndrome.

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Declaration of Interest

The authors report no declarations of interest.

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Table 1

Description of articles (N = 37).

Characteristic	<i>n</i>	Percentage of Studies
Journal Title		
<i>Augmentative and Alternative Communication</i>	7	18.9%
<i>Research in Developmental Disabilities</i>	5	13.5%
<i>Journal of Applied Behaviour Analysis</i>	4	10.8%
<i>Journal of Speech, Language, and Hearing Research</i>	3	8.1%
<i>American Journal on Mental Retardation</i>	2	5.4%
<i>Behavioural Interventions</i>	2	5.4%
<i>Down Syndrome Research and Practice</i>	2	5.4%
<i>Journal of Speech and Hearing Disorders</i>	2	5.4%
<i>American Journal on Intellectual and Developmental Disabilities</i>	1	2.7%
<i>Applied Research in Mental Retardation</i>	1	2.7%
<i>Education and Training in Developmental Disabilities</i>	1	2.7%
<i>Education and Training in Mental Retardation and Developmental Disabilities</i>	1	2.7%
<i>Exceptional Children</i>	1	2.7%
<i>International Journal of Language and Communication Disorders</i>	1	2.7%
<i>Journal of Developmental and Physical Disabilities</i>	1	2.7%
<i>Journal of Intellectual Disability Research</i>	1	2.7%
<i>Topics in Early Childhood Special Education</i>	1	2.7%
Year Published		
1980 – 1989	8	21.6%
1990 – 1999	7	18.9%
2000 – 2009	13	35.1%
2009 – 2014	9	24.3%
Number of Participants (M = 6.86, SD = 14.28)		
1	20	54.1%
2	5	13.5%
3	2	5.4%
4	2	5.4%
5	1	2.7%
> 6	7	18.9%
Total <i>N</i>	37	100.0%

Note: The symbol *n* denotes a number of articles.

Table 2

Participant and study characteristics of intervention studies targeting communication for individuals with Down syndrome.

Authors	Participant Characteristics				Design	Study Characteristics			
	n	n with DS	Mean Age	# Male		Maintenance	Generalization	Integrity	IOA
Bauer & Jones (2014) ¹⁴	5	5	0.85	4	MB Across Behaviours	Y	Y	Y	Y
Bauer et al. (2014) ²⁹	2	2	2.5	2	MB Across Behaviours	Y	Y	Y	Y
Binger & Light (2007) ⁴¹	5	1	4.5	0	MB Across Participants	Y	Y	N	Y
Burgoyne et al. (2012) ²¹	5	57	6.6	28	RCT	N	Y	N	N
	7								
Camarata et al. (2006) ⁴²	6	6	5.7	3	MB Across Participants	N	N	N	Y
Chambers & Rehfeldt (2003) ⁴³	4	1	40	0	AT	N	Y	N	Y
Cottrell et al. (1980) ⁴⁴	1	1	6.6	0	MB Across Behaviours	Y	Y	N	Y
Drash et al. (1989) ⁴⁵	1	15	2.2	9	RCT	N	N	N	Y
	5								
Duker & Michielsen (1983) ⁴⁶	3	1	12	0	MB Across Behaviours	N	Y	N	Y
Feeley et al. (2011) ²⁸	4	4	1.7	1	MB Across Behaviours	Y	Y	Y	Y
Feeley & Jones (2008) ⁴⁷	1	1	3.8	0	MB Across Behaviours	Y	Y	Y	Y
Fey et al. (2006) ¹¹	5	26	4.2	*	RCT	N	N	N	Y
	1								
Gutierrez et al. (2010) ³⁶	2	1	16	1	MB Across Behaviours and AT	N	N	N	Y
Haring et al. (1986) ⁴⁸	3	1	10	0	MB Across Behaviours and Participants	Y	Y	N	Y
Heller et al. (1996) ⁴⁹	4	1	18.3	-	MB Across Participants	N	Y	N	Y
Hemmeter et al. (1996) ²⁷	4	2	6.6	2	MB Across Participants	Y	Y	Y	Y
Iacono & Duncum (1995) ³⁸	1	1	2.7	0	AT	Y	N	Y	Y
Kouri (1988) ⁵⁰	5	1	2.8	0	Reversal	N	N	N	Y
Kroeger & Nelson (2006) ⁵¹	1	1	9	1	Reversal	Y	N	N	Y
LeBlanc et al. (2007) ⁵²	3	1	54	0	MB Across Participants	N	N	Y	Y
Miranda & Dattilo (1987) ⁵³	3	1	11.2	0	MB Across Participants	N	N	N	Y
Poulson (1988) ⁵⁴	3	3	0.44	1	Reversal	N	N	Y	Y
Remington & Clarke (1993a) ⁵⁵	5	3	8.8	1	MB Across Behaviours	N	N	N	Y

Remington & Clarke (1993b) ⁵⁶	6	2	10.9	1	MB Across Behaviours	N	N	N	Y
Rosales & Rehfeldt (2007) ⁵⁷	2	1	34	0	MB Across Participants	Y	N	N	Y
Sepúlveda et al. (2013) ²²	2	20	10.7	11	RCT	N	N	N	N
	0								
Sigafoos et al. (1989) ⁵⁸	3	1	36	1	MB Across Behaviours	N	N	Y	Y
Sigafoos et al. (2009) ⁵⁹	1	1	15	0	AT	N	N	N	Y
Tekin-Iftar (2003) ²⁶	4	1	11.1	1	MB Across Behaviours	Y	Y	Y	Y
Thompson et al. (2007) ⁶⁰	2	1	0.8	0	Reversal	N	Y	N	Y
Tirapelle, & Cipani (1991) ³⁷	2	1	6	1	MB Across Participants	Y	Y	N	Y
Trent et al. (2005) ⁶¹	2	2	6	0	MB Across Participants	Y	N	Y	Y
Valentino et al. (2012) ³⁹	1	1	13.9	0	AT	N	N	N	Y
Warren et al. (1993) ⁶²	5	2	1.8	2	MB Across Behaviours and Participants	N	Y	N	Y
Wright et al. (2013) ⁶³	4	4	2.1	2	MB Across Participants	N	Y	Y	Y
Yoder & Warren (2002) ¹²	3	17	1.8	22	RCT	N	N	N	Y
	9								
Yoder et al. (2014) ¹³	6	35	1.8	-	RCT	Y	N	Y	Y
	4								

“-“ = not reported, Gen =generalization, MB = Multiple Baseline, AT = Alternating Treatments

Table 3

Tailoring to phenotype, intervention characteristics and effectiveness of studies targeting communication intervention in individuals with Down syndrome.

Authors	Tailored to Phenotype		Intervention Characteristics		Intensity Characteristics				PND
	Y/N?	Target Behaviour: Modality, Expressive/receptive, Verbal Operant	Strategies	Opportunity Type	Opportunities /Session	Sessions /Week	Session Duration (min)	Duration of Intervention (sessions)	
Bauer & Jones (2014) ⁶	Y	Voc., exp., echoic and mands	Prompting, Reinforcement	Teacher-led	10	2.5	-	27.75	86%
Bauer et al. (2014) ⁹	Y	Exp., intraverbal	Prompting, Reinforcement	Teacher-led	-	-	10	6.3	100%
Binger & Light (2007) ⁴¹	N	AAC, exp., tacts	Prompting	Learner-directed	30	2	15	29	100%
Burgoyne et al. (2012) ²⁷	Y	Voc., exp. and receptive, undefined	Manualized Reading and Language Intervention	Teacher-led	-	5	40	100	*
Camarata et al. (2006) ⁴²	Y	Voc., exp., undefined	Speech Recasting	Learner-directed	-	-	-	5.5	54%
Chambers & Rehfeldt (2003) ⁴³	N	Signs and AAC, exp., mands	Prompting, Reinforcement	Teacher-led	10	3	36	19	97%
Cottrell et al. (1980) ⁴⁴	N	Voc., exp., intraverbals	Prompting, Reinforcement	Teacher-led	-	6	30	22.67	65%
Drash et al. (1989) ⁴⁵	N	Voc., exp., echoic	Prompting, Reinforcement	Teacher-led	-	12	10	24	*
Duker & Michielsen (1983) ⁴⁶	N	Signs, exp., tacts	Prompting, Reinforcement	Teacher-led	-	3	30	17.3	72%
Feeley et al. (2011) ⁵	Y	Voc., exp., echoic and mands	Prompting, Reinforcement	Teacher-led	5.5	3	15	8.71	76%
Feeley & Jones (2008) ⁸	Y	Voc., exp., tacts	Prompting, Reinforcement	Teacher-led	10	10.5	-	8.33	76%
Fey et al. (2006) ²⁸	N	Voc., exp., mands and tacts	Milieu Teaching	Learner-directed	-	4	20	80	*
Gutierrez et al. (2010) ⁴⁷	N	AAC, exp., mands	Reinforcement	Teacher-led	-	17	5	9.5	0%
Haring et al. (1986) ⁴⁸	N	Voc., exp., intraverbals	Prompting, Reinforcement	Teacher-led,	3	5	10	19.5	95%

Heller et al. (1996) ⁴⁹	N	AAC, exp., mands	Prompting, Reinforcement	Teacher-led	10	-	-	3	100%
Hemmeter et al. (1996) ⁵⁰	N	Voc., exp., mands	Prompting, Reinforcement	Learner-directed	-	5	20	11.5	88%
Iacono & Duncum (1995) ⁵¹	Y	AAC and voc., exp., tact	Prompting Reinforcement	Learner-directed	-	-	30	6	37.50 %
Kouri (1988) ⁵²	N	Voc. and signs, exp., echoic, intraverbal, mands and tacts	Prompting	Learner-directed	-	-	40	17	65%
Kroeger & Nelson (2006) ⁵³	Y	Voc., exp., echoic, mands, tacts, intraverbals	Prompting, Reinforcement	Combination	-	7	60	14	80%
LeBlanc et al. (2007) ⁵⁴	N	Voc., exp., echoic, mands, tacts, intraverbals	NLP	Learner-directed	10	-	10	15	100%
Mirenda & Dattilo (1987) ⁵⁵	N	AAC, exp., mands	Prompting, Reinforcement	Teacher-led	-	39	10	41	95%
Poulson (1988) ⁵⁶	Y	Voc., exp., undefined	Reinforcement	Learner-directed	-	2.5	12	-	91%
Remington & Clarke (1993a) ⁵⁷	N	Signs, exp., tact	Prompting, Reinforcement	Teacher-led	50	10	-	5	75%
Remington & Clarke (1993b) ⁵⁸	N	Signs, exp., tact	Prompting, Reinforcement	Teacher-led	50	10	-	4.67	100%
Rosales & Rehfeldt (2007) ⁵⁹	N	AAC, exp., mands	Reinforcement	Teacher-led	18	3.5	52.5	7	55%
Sepúlveda et al. (2013) ²⁹	Y	Voc., exp., combination	Morphosyntactic Grammar Intervention	Peer-led	-	2	30	30	*
Sigafoos et al. (1989) ⁶⁰	N	AAC, exp., mands	Prompting, Reinforcement	Teacher-led	-	8	5	-	100%
Sigafoos et al. (2009) ⁶¹	N	Voc., exp., mands, tacts	Prompting, Reinforcement	Teacher-led	15.5	3	22.5	-	99%
Tekin-Iftar (2003) ⁶²	N	Voc., exp., tact	Prompting, Reinforcement	Peer-led	15	7	-	17	100%
Thompson et al. (2007) ⁶³	N	Sign, exp., mands	Prompting, Reinforcement	Teacher-led	-	10	5	100	70%
Tirapelle & Cipani (1991) ⁶⁴	N	Voc., sign, exp., mands	Prompting, Reinforcement	Teacher-led	1	7	-	30	47%
Trent et al. (2005) ⁶⁵	Y	Voc., exp., combination	Mirroring and responding	Peer-led	-	-	5	20	24%

Valentino et al. (2012) ⁶⁶	N	Voc., exp., Intraverbals	Prompting, Reinforcement	Teacher-led	36	-	-	33	48%
Warren et al. (1993) ⁶⁷	N	Voc., exp., echoic, intraverbal, mand, tact	Milieu Teaching	Learner-directed	-	4	25	62	91%
Wright et al. (2013) ⁶⁸	Y	Voc., signs, exp., echoic, intraverbal, mand, tact	Milieu Teaching	Learner-directed	-	2	25	20	77%
Yoder & Warren (2002) ⁶⁹	N	Voc., exp., mands, tacts	Milieu Teaching	Learner-directed	-	3.5	20	-	*
Yoder et al. (2014) ¹⁷	N	Voc., exp., mands, tacts	Milieu Teaching	Learner-directed	-	3	60	108	*

Voc. = vocalizations, Exp. = expressive, “-“ = not reported, “*” = PND not calculable

Table 4

Percentage non-overlapping and treatment intensity variables data by treatment type.

Intervention Strategy	<i>n</i>	PND	% Reporting Session Duration	Session Duration (minutes)	% Reporting Opportunities per Session	Opportunities /Session	% Reporting Sessions per week	Sessions /week	% Reporting Total Intervention Duration	Total Intervention Duration (sessions)
Prompting and Reinforcement	2		73.07%		48.1 %		81.5 %		88.0 %	
Mean	7	78.0 %		22		18.86		8		20.05
SD		24.6 %		16.21		16.28		7.92		19.87
Morphosyntactic	1	**		30		*		2		28
Manualized Reading and Language Intervention	1	**		40		*		5		100
Speech Recasting	1	54.0 %		*		*		*		5.5
Milieu Teaching	5		100 %		0.0 %		100 %		80.0 %	
Mean	2	84.0 %		30		*		3.33		67.5
SD		9.9 %		16.95		*		0.84		37.89
Mirroring and Responding	1	24.0 %		5		*		8		20
NLP	1	100 %		10		10		*		15

* not reported **not calculable

Table 5

Intensity characteristics of included studies.

Characteristic	<i>n</i>	Percent Reporting	Range	Mean	SD
Session Duration (minutes)	28	75.68%	5 – 60	23.3	16
Opportunities/Session	14	37.84%	1 – 50	18.3	15.9
Sessions per week	29	78.38%	2 – 39	6.9	7.2
Total Intervention Duration (sessions)	33	89.19%	3 – 108	27.9	29